



Anastomosing Hemangioma: Potential Differential Diagnosis for Incidental Vascular Lesion

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Case Report

The anastomosing hemangioma is an extremely rare benign vascular tumor first described in 2009 by Montgomery and Epstein, previously classified as variants of hemangioma or well-differentiated angiosarcoma [1,2]. Initially conceived as the genitourinary tract characteristic (kidney, testicle and ovary). In recent years, various authors have reported cases in the adrenal gland, liver, colon, small intestine, and mesentery [3,4]. The incidence of anastomosing hemangioma is unknown, although there are hardly any isolated case series described. Anastomosing hemangiomas show a slight predilection for women (1.38: 1) [2], with 53 years of the mean age at diagnosis. With an average size of 2 cm, they are usually unilateral, occasionally occurring bilateral and often multifocal [5]. Although the etiopathogenesis is unknown, there are several cases described in the literature associated with clear cell renal carcinoma and also with end-stage renal disease, especially related to acquired cystic kidney disease [5,6]. Likewise, somatic mutations in the alpha-14 guanine nucleotide binding protein subunit, encoded by the GNA14 gene, appear to be common among a variety of sporadic and congenital lesions of the small vessels, including anastomosing hemangioma [7]. We present a case of a nodular lesion in the right renal hilum without a clear preoperative diagnosis with a pathological report of anastomosing hemangioma.

A 67-year-old male with an allergy to diclofenac and moxifloxacin and a medical history of hypertension, chronic obstructive pulmonary disease, hypercholesterolemia, and mild mitral regurgitation. On treatment with candesartan/hydrochlorothiazide, ipratropium bromide, atorvastatin, omeprazole, silodosin, mometasone and formoterol/budesonide. He was visited for presenting repeated episodes of diffuse abdominal pain accompanied by loss of consciousness during last 3 years. Not another associated symptoms or signs. Exploration without findings of interest. Analytically without relevant alterations. Electrocardiogram (ECG), ergometry and baseline and post-stress transthoracic echocardiogram without evidence of myocardial ischemia or other indicative risk data. In fibrocolonoscopy, 6 polyps were resected, all of them negative for malignancy on pathological examination. Abdominal CT shows a 40 mm × 31 mm nodular lesion in the right renal hilum, hyper-uptake with internal areas of degeneration, between the confluence of the inferior vena cava and the right renal vein. The findings suggest an extraadrenal paraganglioma (Figure 1). The study was completed with a scintigraphy and a SPECT-CT, without appreciating uptake in the previously identified hilar nodular lesion.

In view of the diagnostic doubt and after presenting the case to the Multidisciplinary Committee, a surgical intervention was decided, accessing by means of a right subcostal laparotomy. We found a nodular lesion of approximately 4 cm × 3 cm inferior to the right renal vein, closed with the right renal vein, inferior vena cava and right gonadal vein with each other, with a vascular appearance macroscopic characteristic (Figure 2).

Under the clinical suspicion of vena cava angiosarcoma, it is individualized and excised. Immediate postoperative period in the Intensive Care Unit with good evolution and hemodynamic stability, being discharged to the ward 24 h after surgery. He was discharged from hospital on the seventh postoperative day (Clavien 0). The macroscopic examination showed a nodular lesion of 3 cm × 2 cm × 2 cm with a violaceous surface and a cystic area with solid foci. Microscopically, a well-defined, non-encapsulated nodular lesion is observed, focally infiltrating the adjacent adipose tissue, consisting of cell proliferation that forms capillary-like anastomosing sinusoidal vascular channels, lined by a single layer of cells without significant atypia. The immunohistochemical study determined positivity for the CD31 and CD34 markers as well as the presence of myoepithelial cells, with a ki67 proliferation index of less than 1%. The final histological report was an anastomosing

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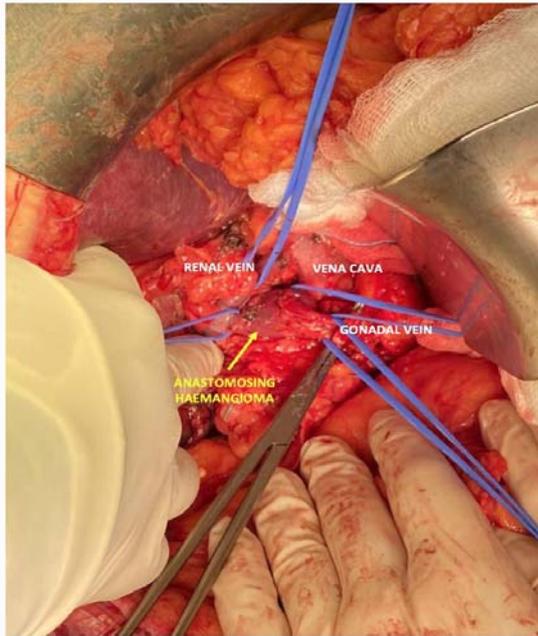


Figure 1: Extraadrenal paraganglioma.

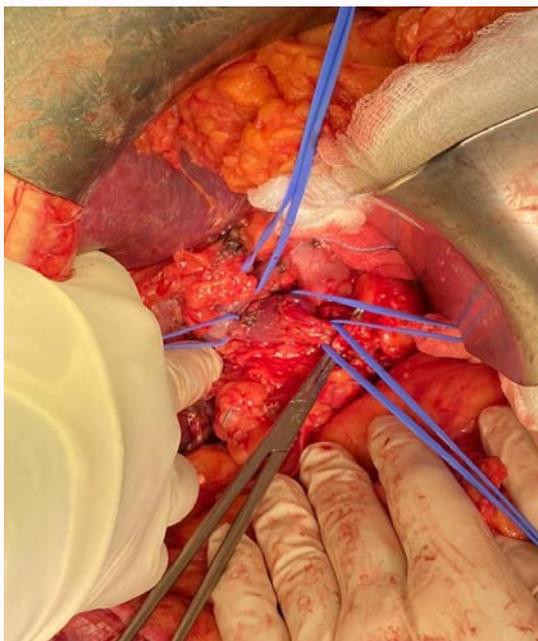


Figure 2: Right renal vein.

hemangioma.

With this diagnosis, the patient did not require subsequent adjuvant treatment, and did not present local recurrence or signs of disseminated disease at the 18-month post-intervention follow-up.

A wide spectrum of symptoms has been described depending on where it is located, however the anastomosing hemangioma is a characteristically indolent neoplasm [5]. Most of the time it is an incidental finding on imaging tests, detected on abdominal CT as a circumscribed, hyperdense and heterogeneous with nodular enhancement in both arterial and venous phases after intravenous contrast-enhanced CT, reported as paraganglioma or metastatic

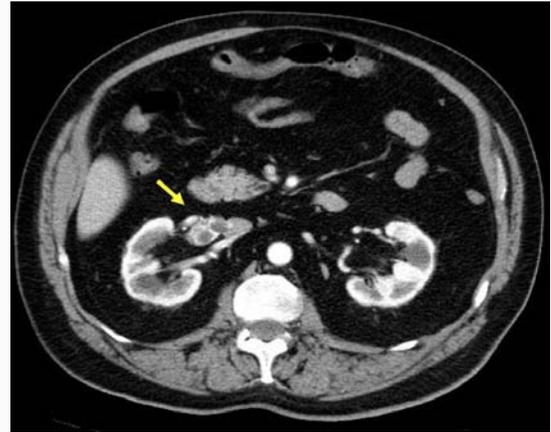


Figure 3: CT.

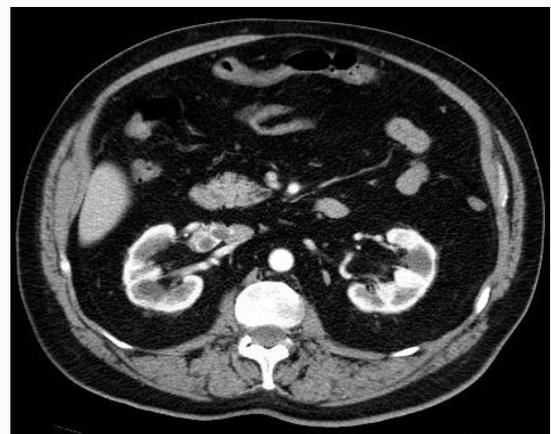


Figure 4: CT.

adenopathy [2]. Ultrasound-guided FNA (Fine-Needle Aspiration) can help to get the diagnosis. Due to the differential diagnosis with other vascular tumors, many patients obtain the definitive diagnosis after surgery. Sinusoidal anastomosing capillaries [3] with tack endothelial cells and positivity for markers CD31 and CD345, [8] are some of their differentiating pathological characteristics. Usually the treatment of anastomosing hemangioma is surgical, and a nephrectomy is the most frequently performed due to its usual location [1]. A pathological diagnosis with FNA previously has made of the observation and the embolization, a therapeutic option [5]. The prognosis is good and local recurrence or distant metastases have not been described in the literature [3].

In conclusion, the anastomosing hemangioma, although rare and infrequent, must be considered as a potential differential diagnosis when an incidental vascular lesion is found.

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